Journal of Pediatric Surgery Case Reports 52 (2020) 101334

Contents lists available at ScienceDirect

Journal of Pediatric Surgery Case Reports

journal homepage: http://www.elsevier.com/locate/epsc

Stump appendicitis in two adolescent boys: Rare presentations and alternative approaches in pediatrics

Sofia Ferreira de Lima [a,b,*], Joana Patena Forte [b], Mafalda de Castro [b], Sofia Morão [a], Rui Alves [a]

[a] Hospital Dona Estefânia, Centro Hospitalar Lisboa Central, Portugal
[b] Hospital Prof. Doutor Fernando Fonseca, Portugal

ARTICLE INFO

Keywords:
Stump appendicitis
Adolescents
Recurrent appendicitis

ABSTRACT

Stump appendicitis is a rare complication that results from reinflammation of the residual part of the appendix, after incomplete appendectomy. Factors that may contribute to inadequate identification of appendicular base are subserosal or retrocecal position, extensive inflammation or inadequate surgical exposure. The length of the appendiceal stump after ligation is likely a major predisposing factor. The presentation of stump appendicitis is similar to acute appendicitis, but this diagnosis is often dismissed because of the surgical history. Computed tomography and ultrasonography are the imaging modalities of choice. The usual recommended treatment for stump appendicitis is completion appendectomy. Stump appendicitis must be considered in the differential diagnosis of acute abdomen despite the patient's appendectomy history. A delayed diagnosis may lead to an increase in morbidity.

We present two cases of stump appendicitis in adolescents after laparoscopic appendectomies, one being a case of recurrent stump appendicitis. We report two alternative therapeutic approaches: interval appendectomy and non-operative management. We conclude that these alternative approaches may be suitable for some patients with stump appendicitis. A review of the pertinent literature was done.

1. Introduction

Appendicitis is the most common surgical emergency in children [1]. The overall lifetime risk is estimated to be around 8%, and it most commonly occurs in the second and third decades of life [1,2]. Stump appendicitis is a rare complication that results from reinflammation of the residual part of the appendix, after incomplete appendectomy [3]. This may still occur from months to decades later with peritonitis and other complications as perforation, abscess formation, appendicocutaneous fistula and sepsis [4,5].

The incidence of stump appendicitis is reportedly 1 in 50,000 appendectomies [6]. From the first report in 1945 until 2018, 164 cases of stump appendicitis have been documented in literature [7]. According to Tang as of 2011 there were only 10 pediatric cases reported in the English literature [4]. However, the exact prevalence is not known and likely underreported as it may imply inadequate surgical technique [8].

Stump appendicitis may occur owing to a long appendiceal residue that resulted from inadequate identification of the appendiceal-cecal junction during an appendectomy [4]. Factors that may contribute to inadequate identification of appendicular base are multiple adhesions, subserosal or retrocecal appendix, extensive inflammation or inadequate surgical exposure [4,9].

The presentation of stump appendicitis is often the same as acute appendicitis, but this diagnosis is often dismissed because of the surgical history [4]. Cases of recurrent appendicitis were reported as stump appendicitis but also as the inflammation of the appendix tip [2]. It was reported as a complication of incidental appendectomy during procedure for duodenal obstruction as an infant [10]. Also reported was a case of recurrence status post two prior appendectomies [3]. Herein we present two cases of stump appendicitis in adolescents and two alternative treatments: interval appendectomy and a nonoperative approach.

2. Case report

2.1. Case 1

A 15 year-old male presented to our pediatric emergency department with a 24 h history of abdominal pain in the right lower quadrant. There was no fever, vomiting or diarrhea complaints. The patient had a prior history of laparoscopic appendectomy for perforated gangrenous

* Corresponding author. Hospital Dona Estefânia, Centro Hospitalar Lisboa Central, Portugal.
E-mail address: sofia.lima@hff.min-saude.pt (S. Ferreira de Lima).

https://doi.org/10.1016/j.epsc.2019.101334
Received 11 October 2019; Accepted 3 November 2019
Available online 9 November 2019
2213-5766/© 2019 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

appendicitis, 9 months before. Since that procedure, he had experienced recurrent episodes of right lower quadrant abdominal pain that were less severe than the current episode. Abdominal examination revealed right lower quadrant tenderness. Further investigation showed 9100 cells/mm^3 leucocyte with 37,9% neutrophil and C-reactive protein <0,29 mg/dl. An ultrasonography (US) was performed and it reported "probable inflammation of appendicular stump, painful and non-compressible at sonopalpation, measuring 21 mm of extension and 11 mm of transverse diameter" (Fig. 1). It was decided to observe the patient for further assessment 24 h later. The right lower quadrant pain persisted with tenderness and he showed leukocytosis (14700 cells/mm^3 with 70,9% neutrophil) and negative C-reactive protein on blood tests. The US was similar to the previous. He showed pain relief after analgesic treatment, so he was discharged and scheduled for a reevaluation 24h later. On the next day he had no abdominal pain. He presented again to the emergency department in three other episodes 3 and 15 months later. He had abdominal right lower quadrant pain associated with diarrhea or sporadic vomits which resolved spontaneously.

An interval laparoscopic surgery was performed and a 2 cm long appendicular stump was identified and resected. Histopathologic examination of the operative specimen showed mucosal lymphoid tissue hyperplasia and submucosal and muscular obliterative fibrosis (Fig. 2). The postoperative period was uneventful. He had no other visits to emergency department or general practitioner for abdominal pain in the 5 years that followed.

2.2. Case 2

A 15-year-old male was admitted to the emergency department with a 12 h history of abdominal pain in the right lower quadrant, nausea, fever (38,5 °C) and bloody stools. He had undergone a laparoscopic appendectomy 4 days before due to a simple appendicitis. He had an uneventful recovery and had been discharged asymptomatic, after a 2-day period of cefoxitin treatment. The histopathologic report confirmed acute appendicitis with reference to a hemorrhagic lumen. Abdominal examination showed surgical wounds with no inflammatory signs and tenderness in right lower quadrant, with no rebound. He had leukocytosis (leucocytes 22,000 cells/mm^3, neutrophils 88%) and increased C-Reactive Protein of 26 mg/L on blood tests. An abdominal US revealed an appendicular stump with thickened walls, painful at sonopalpation, with acute inflammatory changes, measuring around 9 mm of extension and 7 mm of transverse diameter; a diffuse edema of large bowel, most significant on the caecum and fluid collected near the

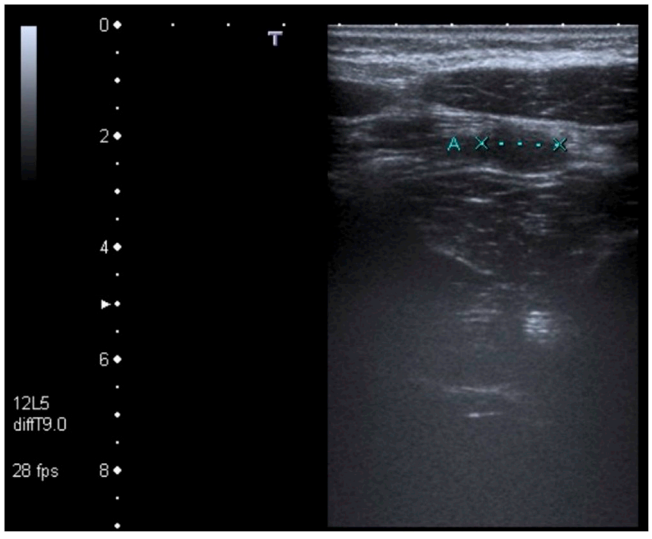

Fig. 1. Longitudinal ultrasound scan showing tubular blind-ended structure measuring 21 mm of extension.

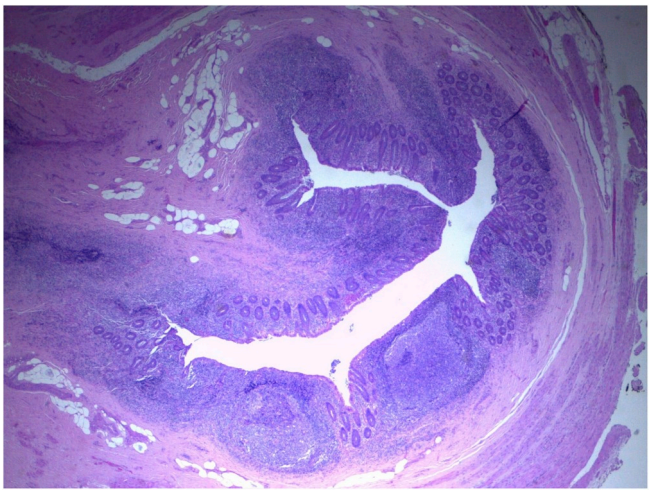

Fig. 2. Photograph of appendicular stump histopathological examination (H&E).

appendicular stump were also observed (Figs. 3 and 4). A conservative approached was adopted with antibiotic treatment. The stool cultures for *Salmonella, Shigella, Campylobacter, Yersinia* were negative; stool ova and parasites tests, *Clostridium difficille* and *Cryptosporidium* were also negative. At the 5th day of Cefotaxime, gentamicin and metronidazole course an ultrasonography was performed. It showed no inflammatory changes on large bowel, but signs of stump appendicitis persisted, as well as some evolving fluid and local mesenteritis. Blood tests showed normal white cell count (6760/mm3, Neutrophils 63,6%) and decreased C-reactive protein (11,7 mg/L).

The patient recovered well and was discharged from hospital after an 8- day antibiotic course. Six months following discharge he remains asymptomatic.

3. Discussion

Stump appendicitis is a rare, long-term complication of appendectomy. Stump appendicitis is an underreported and poorly defined condition. It occurs across all age groups with a mean age of presentation of 35,8 ± 17 (range 2–75 years). The reported interval from original appendectomy to stump appendicitis ranged from 4 days to 50 years [7, 11].

Many are of the belief that the incidence of stump appendicitis may be higher with laparoscopic appendectomy. The arguments for this theory would be the smaller field of vision, lack of three-dimensional perspective, and the absence of tactile feedback [12]. However, in the

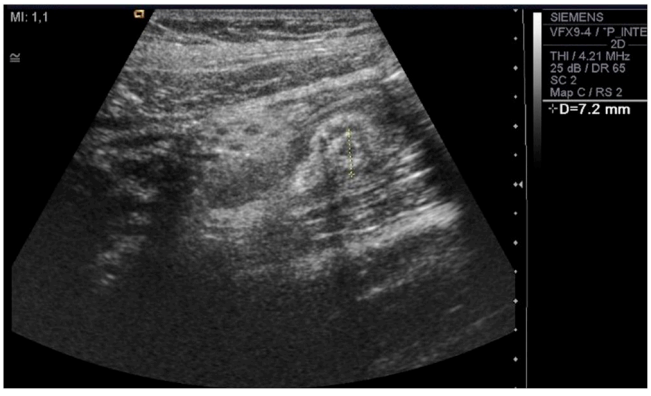

Fig. 3. Transverse (short axis) ultrasound scan showing stump appendicitis with 7,2 mm in diameter.

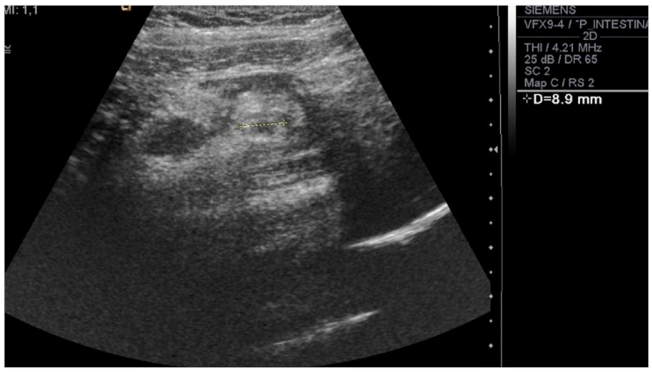

Fig. 4. Longitudinal ultrasound scan showing a tubular blind-ended structure and surrounding fluid.

literature there is no statistically significant increase in incidence of recurrent appendicitis after laparoscopic vs open appendectomy [7,13]. In fact, Manatakis et al. found in their literature review, that of the 160 cases reported, 38% occurred in laparoscopic surgeries whereas 59% occurred in open laparotomies. The length of the appendiceal stump after ligation is likely a major predisposing factor. According to the review by Subramanian et al. there were no cases of stump appendicitis where the appendiceal stump was less than 0,5 cm in length. This stump extension is considered large enough for a fecalith to became impacted and cause obstruction and inflammation [13]. Regardless of the method to remove the appendix it is imperative to clearly recognize appendiceal-cecal junction. A laparoscopic appendiceal "critical view of safety", equivalent to that in cholecystectomy was proposed, where the appendix, the terminal ileum and the taenia libera form a triangle [13].

In case 1, there seems to be a condition of recurrent stump appendicitis [1]. Although the natural history of untreated appendicitis may be perforation and abscess development, this course may not be linear. Spontaneous resolution without specific treatment can occur as is seen in cases of relapsing or chronic appendicitis. Four studies have reported on patients with a clinical diagnosis of appendicitis verified by CT or ultrasound who were not operated on since their symptoms resolved without surgery [14]. Similarly, in case 1, symptoms resolved without antibiotic treatment but recurred frequently, so an interval stump appendectomy was sought.

Imaging studies such as abdominal US, computerized tomography (CT), fistulography, contrast enema and colonoscopy all have been used successfully to diagnose stump appendicitis [4].

Ultrasonography can be useful in identifying inflammatory changes. An enlarged appendix with abnormal cutoff diameter >6 mm suggests stump appendicitis. Additionally, compressibility of the stump appendix relative to adjacent bowel is assessed by graded compression or sonopalpation with the ultrasound transducer [8]. One literature review of 164 reported cases of stump appendicitis concluded that CT, US and their combination had similar rates of high suspicion or correct diagnosis and did not differ with patient's gender or age [7]. The abdominal CT is reported as the gold standard for the diagnosis. According to some authors, CT should be considered as the initial diagnostic study in patients with lower quadrant symptoms after appendectomy or at least would be preferable in ambiguous cases as it provide more details [7,12]. The choice between US and CT in a stump appendicitis suspicion is largely dependent on institutional preference and available expertise [8].

Regarding systemic inflammatory markers, leukocytosis is usually present on admission, although white blood cell count may be within normal range in up to 20% of patients, as was in case 1. C-reactive protein levels were not reported in most studies [7].

Having a previous history of appendectomy may lead to a delay in obtaining imaging in the patients presenting with vague symptoms. It is only when patients present toxic or with peritoneal signs that imaging is performed. This may justify the high rate of complications that are seen in many cases of recurrent appendicitis. The rate of perforation in stump appendicitis is 40–70% compared to 17% in acute appendicitis reported in literature [8,10,12].

A treatment of choice for stump appendicitis is completion appendectomy. However, there were nine reported cases that were successfully treated with nonoperative treatment, as was our case 2 [7,8,11]. There are growing evidence that a conservative treatment with antibiotics for acute appendicitis is safe, especially when there is no evidence of fecalith or other complications as perforation [1]. Non-operative management for stump appendicitis may be suitable for some patients. Close follow-up in these patients is warranted, as concern for recurrent stump appendicitis may approach recurrence rates in non-operatively managed appendicitis [8].

Surgical treatment may include open or laparoscopic completion appendectomy [4,11]. Moreover, it appears that the laparoscopic approach is technically feasible in experienced hands, even after an initial open procedure, with an acceptable conversion rate of 18,5%. Ileocecal resection was performed in more than 15% of patients in a review [15]. This more extensive surgery is usually not necessary if appendicular stump is easily identifiable and caecum wall is not severely compromised by the inflammatory process [16].

Stump appendicitis must be considered in the differential diagnosis of acute abdomen despite patient's history of appendectomy. A delayed diagnosis may lead to delays in treatment and subsequently to an increase in morbidity [6].

4. Conclusion

Recurrent appendicitis is a rare and challenging diagnosis to make. A high index of suspicion is required in any patient with right lower quadrant pain and prior appendectomy in order to prevent delayed diagnosis.

The better solution to stump appendicitis is prevention. During appendectomy it is recommended a careful dissection down to the base of the appendix, even if its view may be difficult by inflammatory changes [4]. The appendiceal stump should be less than 0,5 cm as there is no reported case of stump appendicitis with a stump shorter than 0,5 cm [4,8,10]. Nonoperative treatment with intravenous antibiotic therapy may prevent the need for acute surgical intervention and completion appendectomy in some patients.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Funding

No funding or grant support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The following authors have no financial disclosures: SFL, JPF, MC, SM, RA.

References

[1] Peter AD, Wester T. Appendicitis. In: Holcomb GW, Murphy JP, Peter S, Ashcraft KW, editors. Ashcraft's pediatric surgery. Elsevier; 2019. p. 664–78.

[2] Boardman TJ, Musisca NJ. Recurrent appendicitis caused by a retained appendiceal tip: case report. J Emerg Med 2019;57(2):232–4. https://doi.org/10.1016/j.jemermed.2019.03.044.
[3] Kanona H, Al Samaraee A, Nice C, Bhattacharya V. Stump appendicitis: a review. Int J Surg 2010;10(9):425–8. https://doi.org/10.1016/j.ijsu.2012.07.007.
[4] Tang XB, Qu RB, Bai YZ, Wang WL. Stump appendicitis in children. J Pediatr Surg 2011;46(1):233–6. https://doi.org/10.1016/j.jpedsurg.2010.09.049.
[5] Giwa A, Reyes M. Three times a charm…a case of repeat appendicitis status post two prior appendectomies. Am J Emerg Med 2018;36(3):528.e1–2. https://doi.org/10.1016/j.ajem.2017.12.024.
[6] Dikicier E, Altintoprak F, Ozdemir K, Gundogdu K, Uzunoglu MY, Cakmak G, Onuray F, Capoglu R. Stump appendicitis: a retrospective review of 3130 consecutive appendectomy cases. World J Emerg Surg 2018;13. https://doi.org/10.1186/s13017-018-0182-5.
[7] Manatakis DK, Aheimastos V, Antonopoulou M-I, Agalianos C, Tsiaoussis J, Xynos E. Unfinished business: a systematic review of stump appendicitis. World J Surg 2019;43(11):2756–61. https://doi.org/10.1007/s00268-019-05101-z.
[8] Monsomboon A, Nelson BP, Andrus P, Tsung JW. Point-of-care ultrasound diagnosis of stump appendicitis in the emergency department. Ultrasound J 2019;11. https://doi.org/10.1186/s13089-019-0128-5.
[9] Mínguez G, Gonzalo R, Tamargo A, Turienzo E, Mesa A, Vazquez L. An unsuspected clinical condition: appendicitis of appendicular residual, three cases report. Int J Surg Case Rep 2013;4(4):371–4. https://doi.org/10.1016/j.ijscr.2013.01.026.
[10] Yang Y, Clark TR, Phan HH. Stump appendicitis after childhood incidental appendectomy. J Pediatr Surg 2012;47(11):e15–7. https://doi.org/10.1016/j.jpedsurg.2012.06.017.
[11] Leff DR, Sait MR, Hanief M, Salakianathan S, Darzi AW, Vashisht R. Inflammation of the residual appendix stump: a systematic review. Colorectal Dis 2012;14(3):282–93. https://doi.org/10.1111/j.1463-1318.2010.02487.x.
[12] Crocco S, Pederiva F, Zanelli E, Scarpa M, Barbi E, Ventura A. Stump appendicitis seven years after appendectomy. APSP J Case Rep 2013;4(2):33.
[13] Subramanian A, Liang MK. A 60-year literature review of stump appendicitis: the need for a critical view. Am J Surg 2012;203(4):503–7. https://doi.org/10.1016/j.amjsurg.2011.04.009.
[14] Andersson RE. The natural history and traditional management of appendicitis revisited: spontaneous resolution and predominance of prehospital perforations imply that a correct diagnosis is more important than an early diagnosis. World J Surg 2016;31(1):86–92. https://doi.org/10.1007/s00268-006-0056-y.
[15] Roberts KE, Starker LF, Duffy AJ, Bell RL, Bokhari J. Stump appendicitis: a surgeon's dilemma. J Soc Laparoendosc Surg 2011;15(3):373–8. https://doi.org/10.4293/108680811x13125733356954.
[16] Lupinacci RM, Bouchet-Doumenq C, Salepcioglu H, Egels S, Menegaux F, Trésallet C. Stump appendicitis. A diagnostic trap. Clin Res Hepatol Gastroenterol 2014;38(2):e27–9. https://doi.org/10.1016/j.clinre.2012.11.007.